Cost effectiveness of antenatal screening for cystic fibrosis
Cuckle H, Richardson G, Sheldon T, Quirke P

Record Status
This is a critical abstract of an economic evaluation that meets the criteria for inclusion on NHS EED. Each abstract contains a brief summary of the methods, the results and conclusions followed by a detailed critical assessment on the reliability of the study and the conclusions drawn.

Health technology
Sequential carrier testing (mother first then partner - if mother is found to be a carrier) and couple carrier testing (mother and partner both tested together), as strategies for antenatal screening for cystic fibrosis.

Type of intervention
Screening.

Economic study type
Cost-effectiveness analysis.

Study population
Pregnant women and their partners.

Setting
The economic study was carried out in Yorkshire, UK.

Dates to which data relate
The effectiveness analysis was based on studies published between 1990 and 1995. Costs were reported in 1995 prices.

Source of effectiveness data
Effectiveness data were based on a review of previously completed studies and on assumptions.

Modelling
A decision-tree model was used to combine estimates of outcomes and costs.

Outcomes assessed in the review
The carrier detection rates (i.e. proportion of carriers detected by DNA tests) and the rate of uptake of the screening programme were assessed in the review.

Study designs and other criteria for inclusion in the review
The criteria for inclusion in the review were not explicitly stated, although the authors based the assessment of one of their outcome measures on five British pilot studies reported to date.

Sources searched to identify primary studies
Criteria used to ensure the validity of primary studies
Not stated.

Methods used to judge relevance and validity, and for extracting data
Not stated.

Number of primary studies included
One randomised trial was included in the review. The type of the other primary studies included was not specified.

Methods of combining primary studies
Primary studies were combined using a narrative method, with mid-points of the ranges found in studies used for the baseline analysis.

Investigation of differences between primary studies
Not specified.

Results of the review
The carrier detection rate for both screening strategies was taken to be 80% and the uptake for women offered screening was 75%.

Methods used to derive estimates of effectiveness
In the absence of published data, the authors made baseline assumptions regarding carrier status, the uptake rate of prenatal diagnosis and the uptake for partners of carrier women.

Estimates of effectiveness and key assumptions
It was assumed that carrier couples remember their status, but otherwise 20% having missing information and of the remainder, 20% change partners. The uptake rate of prenatal diagnosis was assumed to be 100%; the uptake rate for partners of carrier women was assumed to be 100%.

Measure of benefits used in the economic analysis
The outcome measure used for the economic analysis was the number of affected pregnancies detected.

Direct costs
Costs were analysed from the perspective of the health care system and focussed on the cost of the screening process. This included the cost of information giving, DNA testing, genetic counselling, and prenatal diagnosis. Estimates were made for the cost of screening a hypothetical cohort of one million women. The unit cost for each component was reported, with costs taken from the published literature and from an unpublished pilot study. Costs were expressed in 1995 prices.

Statistical analysis of costs
Not applicable.
Indirect Costs
Not included.

Currency
UK pounds sterling (¥).

Sensitivity analysis
Three two-way simple sensitivity analyses were conducted. Firstly, the carrier detection rate and the rate of uptake of screening were varied and reported for two types of DNA tests. Secondly, the impact on the cost-effectiveness of each strategy was investigated by varying the proportion of those with missing information about their carrier status (as determined in their first pregnancy) and also varying the proportion of those with a new partner. Thirdly, the impact of varying the carrier detection rate and the cost of the DNA test was investigated. Lastly, one-way sensitivity analysis was conducted to consider a 10% reduction in the rate of uptake of pre-diagnosis counselling.

Estimated benefits used in the economic analysis
In both sequential and couple screening programmes, it was estimated that screening one million pregnant women would detect 384 affected pregnancies.

Cost results
The total cost of the sequential screening programme for one million women was estimated at 17,758,000. The total cost of the couple-screening programme was not reported, but can be estimated from the cost-effectiveness ratio to be in the region of 20,352,000.

Synthesis of costs and benefits
The cost of the sequential screening programme was estimated to be 46,000 per affected pregnancy detected. For couple screening, the cost was estimated to be 53,000 per affected pregnancy detected. Incremental analysis was not performed. The cost-per-case detected with both the sequential and couple strategies was shown to fall as the carrier detection rate increased and as the uptake rate increased, although the latter variable had a smaller effect. For both screening strategies, the cost increases steadily according to the proportion with missing information. There is a similar increase in costs with new partners, but only for couple screening. The cost of the DNA test, the largest cost component, was found to be associated with an almost proportionate change in the cost-effectiveness ratio for both strategies. If the uptake of prenatal diagnosis were reduced to 90%, the cost per case detected would rise from 46,000 to 51,000.

Authors’ conclusions
Cost-effectiveness is highly sensitive to the cost of the DNA test and to the carrier detection rate. Couple screening is more expensive than sequential screening. Screening for this disorder is less cost-effective than screening for Down's Syndrome, but not greatly so. There are no economic grounds for not introducing an antenatal screening service for cystic fibrosis into routine NHS practice, although there may be social, ethical and political reasons for not doing so.

CRD COMMENTARY - Selection of comparators
The reason for the choice of comparator was not explicitly stated, although it would appear that couple screening is an alternative method of identifying carrier couples. You, the user of the database, should consider if this method would be relevant to your own setting.

Validity of estimate of measure of benefit:

It is unclear whether a systematic review of the literature on screening for cystic fibrosis was performed, and what
criteria for inclusion were applied to this review. The reliability of the outcome measures used to derive estimates of benefit is therefore uncertain. Differences between the findings of this study and findings from previous studies were considered and analysed.

**Validity of estimate of costs**
The cost of DNA tests and the inclusion of indirect costs were recognised as explanatory factors for differences in the findings of previous studies. In addition, the proportion of affected pregnancies terminated and the inclusion of just one pregnancy per woman screened increased the cost-effectiveness ratio. The cost of training counsellors was not included in the analysis. The cost of providing counselling may therefore have been underestimated and the cost-effectiveness of the screening programmes may therefore have been overestimated.

**Implications of the study**
The present study provides the NHS with information on cost effectiveness needed to inform decision makers concerning the introduction of a screening service for cystic fibrosis. Screening may be better if made available to the general public without active promotion, rather than targeting the antenatal clinic.

**Source of funding**
Department of Health and Yorkshire Regional Health Authority.

**Bibliographic details**
Cuckle H, Richardson G, Sheldon T, Quirke P. Cost effectiveness of antenatal screening for cystic fibrosis. BMJ 1995; 311: 1460-1464

**PubMedID**
8520332

**Original Paper URL**
http://www.bmj.com/content/311/7018/1460

**Other publications of related interest**


**Indexing Status**
Subject indexing assigned by NLM

**MeSH**
Cost-Benefit Analysis; Cystic Fibrosis /economics /genetics /prevention & control; Female; Genetic Counseling /economics; Genetic Services; Genetic Testing /economics; Great Britain; Heterozygote Detection; Humans; Male; Patient Acceptance of Health Care; Pregnancy; Pregnant Women; Prenatal Diagnosis /economics

**AccessionNumber**
21996008013

**Date bibliographic record published**
31/03/2000

**Date abstract record published**
31/03/2000